Left Main Coronary Artery Perforation During Percutaneous Coronary Intervention in a Patient With Noninfectious Aortitis

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Noninfectious aortitis is increasingly recognized as an important cause of aortic aneurysms and dissection. Coronary involvement in noninfectious aortitis has been reported in several case reports and is marked by a high mortality. Here, we describe the case of a 72-year-old patient suffering from aortitis with involvement of the left main coronary artery trunk, who underwent percutaneous coronary intervention (PCI), which was complicated by left coronary artery perforation. Active inflammatory disease of the vessel wall may cause excessive tissue frailty and therefore has to be considered as a risk factor for perforation during PCI.

[Rev Cardiovasc Med. *2014;15(1):66-70 doi:10.3909/ricm0708]*

KEY WORDS

Arteritis • Giant cell arteritis • Coronary perforation • Noninfectious aortitis

Coronary artery perforation is a rare complication of percutaneous coronary intervention (PCI), for which numerous risk factors have been described. However, underlying inflammatory disease, such as that seen in noninfectious aortitis, has not yet been described as a risk factor for this complication. We describe a patient with aortitis with involvement of the left main coronary artery (LMCA) trunk who underwent PCI, which was complicated by left coronary artery perforation following advancement of a stent but before any balloon inflation. To our best knowledge, this is the first such case described, and it highlights the fact that active inflammatory disease of the vessel wall

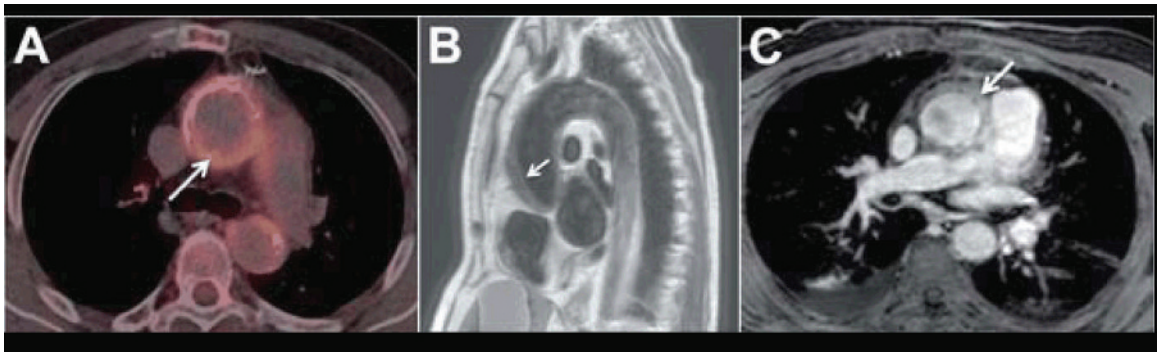

Figure 1. (A) Positron emission tomography-computed tomography scan 3 years prior to admission showing hypermetabolic thickening of the ascending aorta (*arrow*), with a standardized uptake value of 5.9. The ascending aorta is dilated, with a maximal diameter of 41 mm, consistent with inflammation. **(B and C)** Magnetic resonance, T1-weighted high-resolution isotropic volume examination, and T1-weighted images, respectively, on day 2 after admission, showing thickening (13 mm) of the wall of the ascending aorta (*arrows*).

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Case Report

A 72-year-old man with coronary artery disease, moderate aortic valvular disease (both stenosis and regurgitation), peripheral artery disease, hypercholesterolemia, and arterial hypertension, was admitted to our hospital for chest pain at rest, shortness of breath, and fatigue.

Twenty years earlier, he had undergone coronary artery bypass grafting (CABG) of the left internal mammary artery (LIMA) to the left anterior descending coronary artery (LAD). Intraoperatively, aortitis was suspected macroscopically but no biopsy was performed. Later evaluation by magnetic resonance imaging (MRI) and ^{18}F-fluorodeoxyglucose positron emission tomography with computed tomography (CT) confirmed the diagnosis of aortitis (Figure 1A). Results of an autoantibody workup were negative, as results of a Venereal Disease Research Laboratory test, and immunoglobulin G fraction 4 (IgG4) levels were normal. No other systemic symptoms were found, aside from elevated inflammatory markers. Thus, a diagnosis of noninfectious aortitis was made despite the absence of biopsy proof. The patient was started on immunosuppressive therapy with prednisone and methotrexate and subsequent regular clinical and laboratory controls showed marked improvement.

Four months prior to admission, while he was on oral therapy of prednisone, 10 mg/d, and oral methotrexate, 10 mg/wk, a flare of his disease was diagnosed based on elevated inflammatory markers (C-reactive protein [CRP]: 71.6 mg/L [< 10 mg/L]; erythrocyte sedimentation rate [ESR]: 26 mm/h [< 10 mm/h]), in the absence of infection. Cardiac MRI demonstrated increased ascending aorta wall thickening compared with the previous studies with a maximum of 11 mm located circumferentially around the ascending aorta. The patient also reported worsening of his intermittent claudication; Doppler ultrasound studies and CT angiography showed bilateral severe atherosclerotic disease below the knee. Immunosuppressive therapy was stepped-up to prednisone, 20 mg/d, and intramuscular methotrexate, 15 mg/wk. His other medications included lisinopril, amlodipine, and ivabradine. Despite the positive response with regard to inflammatory biomarkers, the patient was reluctant to pursue the prescribed regimen and rapidly switched back to the lower doses of prednisone and methotrexate, resulting in moderately but consistently elevated ESR and CRP values.

On admission, a diagnosis of non–ST-elevation myocardial infarction (NSTEMI) with acute heart failure was made. Laboratory findings confirmed marked inflammation. No source of infection was detected despite thorough investigations. Coronary angiography confirmed the diagnosis, showing a significant stenosis of the ostium of the LMCA, chronic occlusion of the ostium of the LAD, and subtotal distal stenosis of the LIMA bypass anastomosis (Figure 2A). Ventriculography showed diffuse hypokinesis with severe impairment of the left ventricular ejection fraction (30%), and moderate mixed aortic valvular disease. Cardiac MRI revealed persistent signs of aortitis of the ascending aorta with massive thickening and infiltration of the wall (Figures 1B, C). Based on the absence of coronary disease progression other than at the ostium of the LMCA over the past 20 years, we hypothesized that active vasculitis of the aorta surrounding the ostium of the LMCA was responsible for the acute coronary syndrome (ACS). In view of the prohibitive perioperative surgical mortality, estimated according to the Euroscore at 51%, a percutaneous treatment approach was preferred.

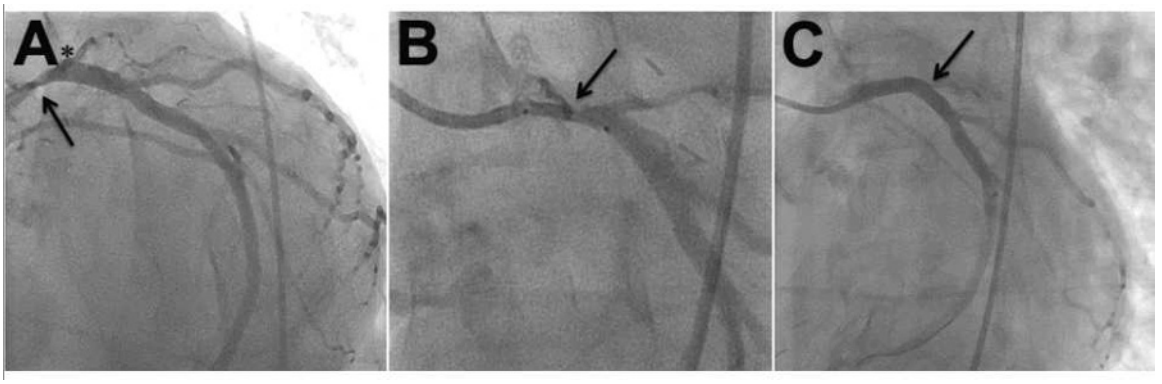

Figure 2. **(A)** Coronary angiography showing a left dominant coronary system with a 70% to 90% ostial stenosis of the left main artery (*arrow*) and chronic occlusion of the ostium of the left anterior descending artery (*asterisk*). **(B)** Coronary angiogram showing perforation of the left main artery following stent advancement, with visible leakage of contrast (*arrow*). **(C)** Coronary angiogram after placement of a drug-eluting stent followed by placement of a covered stent (stent-in-stent) showing good angiographic result with absence of leakage and good vessel patency (*arrow*).

The distal subtotal occlusion of the LIMA bypass anastomosis was treated with a drug-eluting stent (PROMUS Element™; Boston Scientific, Natick, MA), with a good angiographic result. Treatment of the LMCA stenosis was performed using a Launcher® 6F (JL3.5) (Medtronic, Minneapolis, MN) guiding catheter and a S'PORT Extra guide wire (Abbott Vascular, Abbott Park, IL) placed in the circumflex coronary artery, without any attempt to penetrate the chronic occlusion of the ostial LAD. As the PROMUS® 4 × 12 mm stent (Boston Scientific) was advanced, and before any balloon inflation, a perforation of the LMCA was observed (Figure 2B). Heparin was reversed by protamine, and the stent was placed at a maximal pressure of 18 atm. The perforation persisted postinflation with a 4.5-mm noncompliant balloon, thus mandating the placement of a covered stent (JOSTENT GraftMaster 3.5 × 16 mm; Abbott Vascular). Three more balloon inflations with the 4.5-mm noncompliant balloon were performed at a maximum pressure of 20 atm. The final angiogram showed a sealing of the dissection (Figure 2C). His subsequent clinical course was uneventful, serial echocardiography did not show pericardial effusion, the immunosuppressive therapy was intensified by administering intravenous methylprednisolone, 500 mg, over 3 days, and the patient was discharged 10 days after admission on oral prednisone, 40 mg/d, and intramuscular methotrexate, 20 mg/wk. At 3 months, control coronary angiography showed no restenosis and the left ventricular systolic function was improved with an ejection fraction estimated at 45% to 50%.

Ten months later, the patient was readmitted for an NSTEMI with left ventricular failure. Coronary angiography revealed a 70% to 90% intrastent stenosis of the LMCA. Because of the persistently high operative risk, the patient once again underwent PCI. Immediately after drug-eluting stent placement, cardiac arrest with pulseless electrical activity occurred, likely due to acute myocardial failure. No pericardial effusion was detected. Based on the comorbidities of the patient, it was decided not to put him on a left ventricular assist device. All resuscitation efforts failed, and the patient died in the cardiac catheterization laboratory. Autopsy was refused by his family.

Discussion

Noninfectious inflammation of the ascending aorta occurs in the context of large-vessel vasculitis, typically giant cell arteritis (GCA), Takayasu arteritis, and Behçet disease, and less commonly with other systemic and autoimmune diseases such as sarcoidosis, rheumatoid arthritis and spondyloarthritis, granulomatosis with polyangiitis (Wegener), relapsing polychondritis, periaortic and retroperitoneal fibrosis, and IgG4-related aortitis. Idiopathic aortitis may be a subset of GCA or Takayasu arteritis, or occur as a separate disease.[1] Historically, histology has been the gold standard for diagnosis, but some studies indicate low sensitivities, particularly when they include patients with large-vessel GCA.[2] Lately, the possibility of imaging replacing histology, at least in cases with definite findings on clinical examination and imaging, and when histology cannot be obtained, has emerged.[3]

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Approximately 15% of patients with GCA have aortic involvement, and prevalence is even higher in those with Takayasu arteritis.[4] In contrast, isolated thoracic aortitis

in the absence of systemic vasculitis was demonstrated in several recent histopathologic cohorts of surgical specimens of aortic aneurysm resection with a frequency of up to 9%.[5] GCA was by far the most common histologic diagnosis in > 75% of these cases of noninfectious aortitis requiring surgery.[6]

GCA and Takayasu arteritis can be complicated by ACS due to coronary involvement.[7,8] Although the true incidence of coronary arteritis remains undetermined, it is clearly linked to high mortality, as demonstrated by one retrospective study of 90 patients with biopsy-proven GCA, 32 of whom died during follow-up (mean duration 5 y), 13 from myocardial infarction.[9] The suspected mechanisms are accelerated atherosclerosis due to the inflammatory process and acute thrombosis without atherosclerotic changes triggered by high platelet reactivity.[10] The localization of coronary disease is variable. Most reports describe epicardial disease but some also describe intramural disease[11]; however, the incidence of isolated ostial involvement is unknown. In general, aortitis in GCA tends to be continuous and diffuse, possibly accounting for aorto-ostial disease, as seen in our patient, whereas inflammation is typically focal and segmental in smaller arteries.[12]

Thus far, the presence of an underlying inflammatory disease has not been consistently reported as a risk factor for perforation during PCI. Coronary perforation is a rare complication of PCI; it has a 0.43% incidence in unselected populations and is associated with a significant mortality of up to 21.2%.[13] Numerous risk factors have been established, including older age, hypertension, previous CABG, complexity of the lesion, and type of device used.[14] Although the treatment of chronic total occlusions was found to be a strong predictor of coronary perforation in this study, this did not apply in the case of our patient; although he had a chronic total occlusion of the ostium of the LAD, the guide wire was passed only in the circumflex artery and no guide wire placement was attempted in the LAD.

On the other hand, it is well known that aortitis provokes a decrease in aortic distensibility and aortic strain,[15] leading to aneurysm formation and dissection. Nevertheless, several cases of dissection, free rupture, or impending rupture of the thoracic aortic wall without aneurysmal dilation have been reported,[16] demonstrating the underlying tissue frailty. There are no reports in the literature on perforations due to coronary arteritis and only a few have occurred during endovascular treatment in patients suffering from vasculitis. We want to put forward the hypothesis that tissue frailty caused by ongoing aortitis expanding into the coronary ostium has probably precipitated coronary perforation.

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In light of the high complication rate of both coronary surgery (especially if venous bypasses with aortic anastomosis are needed) and PCI, a conservative approach with medical treatment should be carefully evaluated. Partial regression of the LMCA lesion following steroid administration has been reported in a patient with Takayasu aortitis following emergency CABG.[17] In our patient, we were convinced that the association of left dominance, severe LMCA stenosis, and newly depressed left ventricular function mandated LMCA revascularization in addition to the LIMA revascularization because the circumflex was not supplied by this graft. In another case of ACS with underlying Takayasu arteritis, PCI was complicated by three episodes of restenosis, even with the use of drug-eluting stents. Finally, steroid administration provided patency of the stent site, underscoring the importance of treating the underlying inflammatory disease.[18]

Conclusions

We strongly believe that the excessive frailty of the coronary ostium due to ongoing adjacent aortitis led to the observed perforation of the LMCA during stent advancement even before any balloon dilatation. To our best knowledge, this is the first report of coronary perforation during PCI in a patient with suspected proximal coronary arteritis, underscoring the elevated risk of PCI and the need of appropriate immunosuppressive treatment in this situation. ■

The authors report no real or apparent conflicts of interest.

The authors thank Gisella Puga Yung for her help with the preparation of the figures.

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MAIN POINTS

- Noninfectious aortitis is increasingly recognized as an important cause of aortic aneurysms and dissection. Active inflammatory disease of the vessel wall may cause excessive tissue frailty and therefore has to be considered as a risk factor for perforation during percutaneous coronary intervention (PCI). Excessive frailty of the coronary ostium due to ongoing adjacent aortitis can lead to perforation of the left main coronary artery during stent advancement, even before any balloon dilatation.
- Histology has been the gold standard for diagnosis, but some studies indicate low sensitivities, particularly when they include patients with large-vessel giant-cell arteritis. Recently, the possibility of imaging replacing histology, at least in cases with definite findings on clinical examination and imaging, and when histology cannot be obtained, has emerged.
- In light of the high complication rate of both coronary surgery (especially if venous bypasses with aortic anastomosis are needed) and PCI, a conservative approach with medical treatment including appropriate immunosuppressive therapy should be carefully evaluated.